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RUNNING HEAD: Systematic review of speech interventions

Title: A systematic review and classification of interventions for speech sound disorder in preschool children

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Keywords: speech sound disorder, systematic review, Child Talk, intervention

What this paper adds:

- Section 1: *What is already known on this subject.* A wide range of interventions are available for speech and language therapists to use when working with children with speech sound disorder. While some intervention approaches have robust evidence to support them, others do not have evidence or have more limited evidence.
- Section 2: *What this study adds.* This study systematically reviewed the evidence for those interventions which have been tested with children under 6 years of age. A model for classification of intervention studies in speech sound disorder is proposed and the evidence to support interventions within the model provided.
- Section 3: *Clinical implications of this study.* Speech and language therapists will be able to identify at a glance which interventions that have been tested with children under age 6 have evidence to support them. Evidence is varied in strength and intervention studies using more robust research designs are needed to fully test the interventions described in the current literature.

Abstract

Purpose: To systematically review the evidence for interventions for speech sound disorder (SSD) in preschool children and to categorise them within a classification of interventions for SSD.

Method: Relevant search terms were used to identify intervention studies published up to 2012, with the following inclusion criteria: participants were aged between 2 years and 5 years, 11 months; participants exhibited speech, language and communication needs; and a primary outcome measure of speech was used. Studies that met inclusion criteria were quality appraised using the SCED or Pedro-P depending on their methodology. Those which were judged as high quality were classified according to the primary focus of intervention. The review PROSPERO registration number is CRD42013006369.

Results: The final review included 26 studies. Case series was the most common research design. Categorisation to the classification system for interventions showed that cognitive-linguistic and production approaches to intervention were the most frequently reported. The highest graded evidence was for three studies within the auditory-perceptual and integrated categories.

Conclusions: The evidence for intervention for preschool children with SSD is focused on seven out of 11 subcategories of interventions. Although all of the studies included in the review were good quality as defined by quality appraisal checklists, they mostly represented lower graded evidence. Higher graded studies are needed to understand clearly the strength of evidence for different interventions.

Introduction

Speech sound disorder (SSD) is a high prevalence condition in preschool children (Broomfield and Dodd 2004, Eadie *et al.* 2015, McLeod and Harrison, 2009, Shriberg *et al.* 1999). In response to this, a number of interventions have been developed which vary in the method used to achieve change in a child's speech (Baker and McLeod, 2011).

To date, there have been a number of systematic literature reviews that have examined the effectiveness of these interventions for children with SSD across the age range. Some of the reviews were part of a larger and more comprehensive review of speech and language therapy interventions for children with speech and language delay or disorder (Law *et al.* 2003, Law *et al.* 2012, Law *et al.* 2015) while others have focused specifically on speech (Baker and McLeod 2011, Murray *et al.* 2014) or on a specific type of intervention (Lee *et al.* 2009, Lee and Gibbon 2015, McCauley *et al.* 2009, Morgan and Vogel 2008). While those focusing on specific interventions revealed a paucity of studies with sufficient strength to provide categorical support for the approaches (specifically, electro-palatography, Non Speech Oral Motor Exercises, and interventions for Childhood Apraxia of Speech), the results of the more extensive reviews were encouraging. Law *et al.* (2003) included only randomised controlled trials in their review and found convincing support for interventions where the outcome was the child's 'expressive phonology'. Similarly, the review by Law *et al.* (2012) found that out of 57 interventions included in the review, approximately one third (38%) targeted speech. Evidence for most of these interventions was at a moderate level (68%), i.e. tested in either a randomised controlled trial or several quasi-experimental studies, whilst for others the evidence was at an indicative level, i.e. they have good face validity and are widely used by clinicians but have limited research evidence which can be generalised to the population concerned.

Baker and McLeod (2011) included a wider range of study designs in their narrative review of evidence based practice for children with SSD. Samples in these studies included participants with concomitant difficulties such as hearing loss, cleft lip and/or palate, or stuttering and spanned an age range of 1;11 to 10;5. They identified a total of 154 studies which described seven different methods for target selection and 46 different approaches to intervention. While a small number of these interventions had been subject to meta-analysis or included in a randomised controlled trial, the majority had been subject to less rigorous investigations such as quasi experimental or non-experimental case studies. Baker and McLeod concluded that more rigorous experimental design is required to enable the relative benefits of any intervention or approach to be determined.

The interpretation of Baker & McLeod's review in a clinical context is challenging. Authors of differing theories and approaches often provide clear guidance regarding the most appropriate intervention to use with children with differing presentations (for example Dodd & Bradford, 2000. However, without comparisons of the efficacy or effectiveness of one approach over another for the full range of approaches that are available, clinicians are left without clear evidence of the best approach to use. This challenge is well illustrated in the 2006 special edition of *Advances in Speech-Language Pathology* on 'Jarrod', a 7-year-old boy with SSD. This symposium published papers by different authors, who were invited to advocate and describe their own approach to intervention for this child. The different interventions were all well-argued and justified at a theoretical level but not compared with each other and there was no conclusion regarding which approach might be the most effective or efficient.

The recognition that different approaches to intervention may be needed for children with different presentations of SSD has led to a widespread call in the literature for

more detailed assessment and analysis of SSD (McLeod and Baker 2004, Skahan *et al.* 2007, Stackhouse and Wells 1997). In the absence of this, clinicians tend to favour the use of just two or three named approaches, often combined into one eclectic package, presumably with the expectation that one of the elements within the package will target the child's specific needs (Joffe and Pring 2008, McLeod and Baker 2004, Roulstone *et al.* 2012). The approaches named by speech and language therapists as most frequently used often lack detail and are ambiguous in terms of how exactly they are delivered or interpreted. Terms such as 'auditory discrimination', 'meaningful minimal contrast', 'phonological awareness', (Joffe and Pring 2008), 'traditional articulation therapy' and 'minimal pairs', (McLeod and Baker 2004) and 'minimal pairs', 'auditory discrimination' and 'sequencing sounds' (Roulstone *et al.* 2015) being cited as commonly used interventions. It is therefore not clear how far the approaches used frequently by clinicians map onto the approaches described in the intervention literature.

There is a need to systematically appraise the evidence for intervention in SSD and then map that onto the approaches described by clinicians. In this way speech and language therapists with a busy and varied caseload would more easily be able to identify the strength of evidence for interventions which fit with the approach they determine is needed for an individual child.

A model for classification of interventions for SSD

Existing classifications of SSD have focused on the child's aetiology (Shriberg *et al.* 2010), their surface level speech presentation (Dodd, 2005) or their speech processing skills (Stackhouse & Wells, 1997). A useful summary of these approaches is provided in Waring and Knight (2011). While the Dodd classification provides guidance regarding which interventions map onto each identified subtype, this only covers a small number of the

range of interventions that are available, as identified by Baker and McLeod (2011). An alternative approach is to classify interventions and attempt to map this to the kinds of difficulties that children with SSD might experience. This approach has been adopted in descriptions of intervention approaches by Bernthal *et al.* (2012), Rvachew and Brosseau-Lapr  (2012) and Stackhouse and Wells (1997). Typically, interventions have been grouped into the level of processing they are primarily targeting: ‘input’, where the child is required to respond to some auditory stimuli to effect change in their speech; ‘storage’, where the child is asked to reflect on their stored representations of words as a means to challenge existing inaccurate representations; or ‘output’, which require the child to produce speech in response to imitation or some other stimuli.

An extension of this approach was expanded in work carried out by Wren (2005) and was used as the basis for the work carried out in the systematic review reported in this paper. Using a bottom-up approach from the intervention procedures which are available and identified as in use by clinicians (Roulstone and Wren 2001), the model is organised by the area where change is expected to occur in order to facilitate change in speech output. It is hypothetical and proposes one way of organising types of intervention procedures and has changed since the original version described in Wren (2005). As such, it has the capacity to change further and evolve as new intervention procedures and new evidence become available. Nonetheless, it provides an initial framework that is inclusive of the diverse range of intervention procedures that are available to clinicians. Specific approaches are not named in this model but the area where change is expected to occur and which indeed is being targeted in the intervention has been identified and categorised accordingly (Figure 1).

[Figure 1 about here]

The model labels five categories of intervention: environmental, auditory-perceptual, cognitive-linguistic, production and integrated. The environmental approach is distinct from the others in that it encompasses intervention approaches which make use of everyday interactions, rather than specific directed activities, to promote change in a child's speech sound system. This would include procedures sometimes described as 'naturalistic intervention' as well as modelling and recasting of a child's spontaneous productions (Camarata 2010). Auditory perceptual interventions target the child's perceptual skills as a means to induce change in speech output and include activities that aim to increase exposure to the sounds being targeted, as in focused auditory stimulation, and discrimination tasks designed to increase phoneme perception skills (Hodson and Paden 1991, Rvachew and Brousseau-Lapr  2010). Cognitive-linguistic interventions engage the child in higher level processing in which the child's awareness of their speech is consciously addressed and used to promote change, either through confronting a child with their reduced set of contrasts or through increasing awareness of sounds in speech generally. Interventions focusing on production aim to effect change through performance of oro-motor tasks, guidance on phonetic placement or manner, imitation and drills. Integrated interventions are simply those that combine two or more of the other four through profiling of the child's specific needs as in the psycholinguistic approach (Stackhouse and Wells 1997) or combining procedures into a programme of multiple interventions consistent with a Cycles approach to intervention for example (Hodson and Paden 1991).

The model does not reflect decisions around phoneme target selection though undoubtedly, the decisions regarding procedure and target are related for many interventions. Nor does it attempt to link to aetiology. However, the model makes explicit where change is expected to occur as a consequence of intervention. It is anticipated that

this would provide a summary of the current evidence which is more easily accessible to clinicians, and therefore addresses some of the concerns raised in Lancaster *et al.* (2010) regarding the incompatibility of research and clinical work.

Aim of the study

The aim of this study was to systematically review and critically appraise the strength of the evidence for interventions for SSD in preschool children and then categorise those interventions which fulfilled the selection criteria within the model of classifications of interventions for SSD described above. Studies of interest would include children with SSD aged between 2 and 6; use a range of study designs; and measure outcomes in speech. The intention was that this would provide an overview of current evidence for intervention for SSD with preschool children in an easily accessible format which could be quickly be mapped onto individuals' children's needs.

This study was part of a larger review of interventions for children with speech and language impairment in preschool children with no concomitant difficulties (Roulstone *et al.* 2015) within the 'Child Talk' research programme, a series of research studies investigating the evidence base for speech and language therapy intervention for preschool children.

Method

The systematic review was guided by the principles outlined in the Cochrane Collaboration methodology (Higgins and Green 2011), as far as they could be applied to the study methodologies, and built on the review undertaken by Pickstone *et al* (2009). The search strategy described below outlines the larger review carried out for the 'Child Talk' research program and describes how the studies relevant to SSD were identified within this.

The systematic review was registered with PROSPERO (CRD42013006369), an international register of prospective systematic reviews.

Search strategy

The search strategy employed three key elements: development of a comprehensive and relevant list of search terms to ensure that all potentially valid studies in relation to interventions for speech and language impairment without concomitant difficulties were returned; exploration of a suitably broad range of databases to capture as many potentially valid studies as possible, including published, unpublished and conference proceedings; and identification of clear inclusion criteria against which to filter potentially valid studies and provide the dataset for analysis. The authors and co-applicants of the 'Child Talk' programme of research (Roulstone *et al.* 2015) identified a set of search terms based on their previous work in the field (Blackwell *et al.* 2014, Hambly *et al.* 2013, Marshall *et al.* 2011, Pickstone *et al.* 2009, Wren *et al.* 2013) . Further potential search terms were identified from key papers. This expertise was augmented through consultation with information specialists. Through an iterative process of identification and discussion, a list of 90 search terms was determined to provide the most appropriate set to capture potentially valid studies (Appendix 1). The same process was used to select appropriate databases to ensure maximum inclusion of published data, unpublished data and conference proceedings.

In line with Booth and Fry-Smith (2003), the PICO model (Population, Intervention, Comparison, Outcome) guided the development of the inclusion criteria. All research design methodologies were considered and therefore the 'Comparison' element of the PICO model was not used to determine eligibility but recorded during data extraction. For inclusion in the larger 'Child Talk' review, studies had to meet the following requirements:

- Population: At least 80% of the sample were required to be within the age range 2 years to 5 years and 11 months at the start of the intervention or at recruitment; children would be diagnosed or considered 'at risk' of speech and language impairment without concomitant difficulties.
- Intervention: An empirical evaluation of an intervention, including randomised controlled trials, experimental and quasi-experimental studies and case studies which included multiple baseline or other systematic manipulation of the intervention.
- Outcomes: At least one of the primary outcome measures of included studies would address speech, language, communication or interaction (At a later stage, those studies which included primary outcome measures of speech were included in this topic specific review – see below.)

Studies were excluded if:

- They related to children whose speech or language appeared to be developing typically with no evidence to suggest that their language was 'at risk'.
- They related to children whose speech or language delays were associated with other developmental or pervasive conditions such as learning difficulties, autism, cleft palate and cerebral palsy.
- The only outcomes were social or behavioural.

Search procedure

A combination of 'free text' terms with Boolean operators and truncations was used.

Eighteen separate searches were conducted in electronic databases (Appendix 2), to identify

appropriate studies in articles published from the earliest entries of any of the databases until January 2012. Papers were initially reviewed by title and then by abstract.

Reliability

Two of the authors independently reviewed the titles of ten percent of the papers identified from the initial search of the databases to screen for relevance, removing any studies which did not fit the exclusion and inclusion criteria. There was 100% consensus and the remaining 33,000 references were shared between these two authors and papers were excluded at the title level. This process led to the retention of 4,574 papers. The abstract review was undertaken by four members of the research team, with two people for each manuscript (one Speech and Language Therapist and one Psychologist). Where disagreements occurred, discussion took place within the team until consensus was reached. Those papers retained at this stage were then reviewed in their entirety in light of the inclusion and exclusion criteria.

The retained papers were further reduced to those that had interventions which related to SSD. Studies were included at this stage if the intervention described in the research was consistent with the definition: "Work that increases the accuracy of speech production or articulation, often focusing on specific sound(s)". Those studies which focused on phonological awareness skills only and did not relate to speech output were excluded. The remaining papers were then subjected to a quality appraisal.

Quality appraisal

The quality appraisal tools used in this review were selected to be relevant to the research designs used in the included studies. Two tools were used for this purpose: the Physiotherapy Evidence Database quality assessment tool (PEDro-P, Perdices and Tate 2009) had a score range of 0-9 and was used to appraise the methodological quality of randomised

and non-randomised controlled trials and; and Single Case Experimental Design (SCED) had a score range of 0-10 and was used for single case studies (Tate *et al.* 2008). All appraisers undertook and passed training on PEDro-P and SCED (<http://speechbite.com/rating-research-quality/outline-rating-training-program/>). Each article was reviewed by at least two researchers and if disagreement had occurred it was planned to discuss and reach consensus. This process was not required as agreement on the quality assessment was 100 percent. For both tools, a higher score was associated with greater quality of the methodology applied and reported within the study. In line with previous reviews (Camarinos and Marinko 2009, Maher *et al.* 2003), a score of six or over was used to identify studies of acceptable quality which would be retained in the review. These studies were then mapped onto the classification of intervention procedures model described above.

Data extraction and synthesis

The process of synthesis consisted of 2 stages. The first stage extracted the characteristics of the studies relating to country, culture, and language/s of the researchers and participants and to study designs categorised using the National Health and Medical Research Council levels of evidence guidelines (NHMRC 2007). A wide range of study designs were included in the review. This was to acknowledge that those with a lower level of evidence could be developed into trials using higher graded designs in the future.

The second stage extracted information on location and agent of intervention, assessment and outcome measures used, number of treatment sessions and a description of the intervention provided. The description of the intervention was used to map the study to the model of intervention procedures. Specifically, the information provided in the paper which described the procedures (as opposed to targets or the underlying theory) carried out

to effect change in the child's speech sounds was considered to identify the best fit with the categories within the model described in the introduction. Where more than one type of procedure was included in the intervention protocol but only one category was under investigation, the study would be classified under the category which was the best fit for the element of the intervention being investigated. Where a combination of types of procedure had been implemented, these were noted and the study assigned to the 'integrated' category. Table 1 provides a summary of the criteria used to categorise intervention procedures described in each paper.

Subsequently, effect sizes for speech outcomes were calculated where data were available and appropriate. This was undertaken using the Campbell Collaboration effect size calculator (<https://www.campbellcollaboration.org/escalc/html/EffectSizeCalculator-SMD-main.php>). Studies using a within-subject pre-post methodology providing sufficient information were assessed using a second online calculation tool (<http://www.cognitiveflexibility.org/efficientsize/>) and single-subject experimental designs were assessed using Improvement Rate Difference (IRD; Parker *et al.* 2011).

Results

Figure 2 shows the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses, <http://www.prisma-statement.org/> accessed 03/03/2016) flowchart and summary of papers retrieved at each stage of the review. Of the 147 studies matching the inclusion criteria for the Child Talk project as a whole, 55 could be mapped onto the speech theme. Twenty-five of these papers, reporting on 26 studies, demonstrated a sufficient level of quality (i.e. obtained of six or more) when assessed using the PEDro-P or SCED scale.

Of the 30 that did not attain a score of six or more on these measures, 11 were reviewed using PEDro-P and 19 with SCED. The mean average scores on these excluded studies were 4 and 3 respectively (median 4 and 3). The most frequent deficits in the randomised and non-randomised controlled studies were: lack of concealment during group allocation and lack of blinding of the assessor who measured at least one key outcome. In the single case experimental studies, the top three deficits in reporting were: lack of raw data being reported; assessors not being independent of treatment/intervention; and lack of replication either across subjects, therapists or setting.

[Figure 2 about here]

Categorisation of studies and reported outcomes

Of the 26 studies retained for inclusion, 18 were undertaken in the USA, 4 in Canada, 3 in Australia and 1 in the UK. Fifteen of the studies used a case series design and 3 were case studies. A further 3 studies used a randomised controlled trial design and a further 4 used a between groups design. The 26 studies were categorised according to the procedure used in the intervention using the model in figure 3 (see figure 3). It was possible to calculate effect sizes in ten of the studies and to provide a range of the improvement rate difference in single cases for three more. Table 2 details each of the studies in the review and provides summary information on each obtained from the data extraction.

[Figure 3 about here]

Environmental approaches are represented by one study. The study by Yoder *et al.* (2005) was categorised here due to the intervention using recasting and modelling within clinic contexts. This study found no main effect of the broad target recast intervention but did report a positive long term impact on intelligibility for children with low pre-treatment speech accuracy in comparison with standard care.

Within the category of auditory perceptual approaches, the subcategory of phoneme perception approaches was used in three studies (Rvachew 1994, Rvachew *et al.* 2004, Wolfe *et al.* 2003). The children in the Rvachew (1994) study were randomly allocated to three groups and these children were given listening tasks focused on treatment of misarticulated versions of target words. Rvachew *et al.* (2004) used training in phonemic perception, letter recognition, letter-sound association and onset-rime matching. Both studies found a positive effect of the intervention. In contrast, Wolfe, Presley and Mesaris (2003) compared sound identification training plus production training with production only training and found no difference between the two groups except for sounds which were poorly identified prior to intervention. None of the studies in the review were classified under the focused auditory stimulation subcategory.

Cognitive-linguistic approaches were the most commonly reported interventions within the studies in the review. These studies focused on three subcategories of intervention: 'meaningful minimal contrast' approaches, 'complexity' approaches and 'metalinguistic approaches'. Three studies focused on meaningful minimal contrast (Baker and McLeod 2004, Dodd and Iacono 1989, Robb *et al.* 1999) and a further six studies (from five papers) form the evidence base for (Gierut 1989, 1990, Gierut and Champion 1999, Gierut *et al.* 1996) and against complexity approaches (Rvachew and Nowak 2001). These studies have small samples but suggest a positive impact of the interventions on the children, with one exception where change to the target of intervention was not observed (Gierut and Champion 1999). No studies were included in the review under the category of metalinguistic approaches.

Studies within the review which came under the category of production were identified within the subcategories of 'oro-motor speech exercises', 'guidance on phonetic

placement/manner' and 'imitations and drill'. No studies were categorised under 'oro-motor speech exercises' or 'guidance on phonetic placement/manner'. The seven studies within the 'imitations and drill' subcategory all worked on increasing the complexity of articulation in graded steps such as breaking words into constituent sounds and subsequently recombining to form the word (Forrest and Elbert 2001, Forrest *et al.* 2000, Gierut 1996, Gierut and Champion 1999, 2001, Gierut and Morrisette 1996, Winner and Elbert 1988). Five of these studies showed an improvement in the intervention group (Forrest and Elbert 2001, Forrest *et al.* 2000, Gierut and Champion 2000, 2001, Gierut and Morrisette 1996), while in two studies there was no statistical impact of the intervention on the child's speech output (Gierut 1996, Winner and Elbert 1988). It is important to note, however, that the purpose of the intervention in Winner and Elbert's study was to investigate the impact of administering repeated probes during intervention with the intention that a desired outcome would be no change in performance on the probe measure, indicating that this approach can continue to be used in future trials of intervention for SSD.

'Integrated' approaches to intervention were represented by studies within the subcategories of 'combined' approaches and 'unspecified'. Combined approaches were adopted in four studies included in the review (Almost and Rosenbaum 1998, Hart and Gonzalez 2010, McIntosh and Dodd 2008, Saben and Ingham 1991). The studies used a combination of activities and strategies as interventions, described as being targeted at the individual child's needs or as routine one-to-one therapy. The studies provide mixed evidence for this approach: Almost and Rosenbaum (1998) showed a positive effect of active therapy in a group study while the remaining three studies reported case studies with varying patterns of response from individuals. Unspecified approaches were used in the

Glogowska *et al.* (2000) study where no differences overall were found on the phonology score between control children and those receiving standard treatment. However, on a secondary outcome, a significantly greater proportion of children receiving treatment compared to the watchful waiting group improved their phonology such that they no longer satisfied the original phonology eligibility criteria for the trial.

Delivery of intervention

All studies included in the review used interventions that were delivered by speech and language therapists. Several studies did not provide information on the number and length of intervention sessions, however where they did, the range was from three to 67 sessions lasting between 30 and 60 minutes.

Assessment measures used

Speech measurement in the reviewed studies was carried out for one or more of three purposes: to confirm eligibility for participation in the study; to identify targets for intervention; or to measure change in response to intervention (outcome measure). Three studies, all within the subcategory of phoneme perception approaches, also measured change in speech perception (Wolfe *et al.* 2003, Rvachew *et al.* 2004, Rvachew 1994). Speech output was collected using published assessments (Hart and Gonzalez 2010, McIntosh and Dodd 2008, Rvachew and Nowak 2001), confrontation picture naming tasks devised for the study (Saben and Ingham 1991, Winner and Elbert 1988), and spontaneous continuous speech samples (Dodd and Iacono 1989, Hart and Gonzalez 2010, Saben and Ingham 1991, Rvachew 1994, Rvachew *et al.* 2004, Winner and Elbert 1988, Yoder *et al.*, 2005). In all studies, reliability of the transcriptions was reported using point-to-point agreement for two transcribers, from between 20 to 100 percent of data collected. Some studies used a combination of two or three approaches to collecting speech samples.

Several studies also used picture naming as part of a probe testing protocol (Baker and McLeod 2004, Forrest *et al.* 2000, Forrest and Elbert 2001, Gierut 1996, 1990, 1989, Gierut *et al.* 1996, Gierut and Champion 2000, 1999, Robb *et al.* 1999, Saben and Ingham 1991, Wolfe *et al.* 2003).

In terms of analysis of the speech samples collected, those studies which included published assessments within their assessment protocol typically used the analysis procedures which accompanied those tools. These included process analysis (Assessment of Phonological Processes-Revised, Hodson, 1986, 2004), phonemic or phonetic inventories, (Productive Phonological Knowledge Profile, Gierut *et al.* 1987), percentage phonemes/consonants/vowels correct (Diagnostic Evaluation of Articulation and Phonology, Dodd *et al.* 2002, Shriberg and Kwiatkowski 1982), and accuracy of production (Goldman-Fristoe Test of Articulation, Goldman and Fristoe 2000). Where spontaneous speech samples, confrontation picture naming or probe lists were used, a number of analyses were carried out, as detailed in table 2.

[table 2 about here]

Discussion

This systematic review of the literature has considered the evidence for a range of interventions for preschool children with SSD within a model in which interventions were classified based on the nature of the procedures used to effect change. In total, 55 papers were identified based on clearly defined search criteria. Following quality appraisal, 25 papers reporting 26 studies were appraised as robust enough to be included in the final review. These 26 studies were then mapped onto the model of interventions according to the description of the procedures within each paper.

Description of the review

While some previous reviews have limited their enquiry to children with phonological problems only (Baker and McLeod 2011), this review included any study which targeted increased accuracy of speech production or articulation, encompassing both phonological and speech motor interventions. This was important given the aim of synthesizing the evidence for clinicians who will be faced with a broad spectrum of children with SSD in practice (Broomfield and Dodd 2004, Shriberg *et al.* 2005).

The review included a range of research designs and did not limit itself to RCTs, though most were at level III of the NHMRC Evidence Hierarchy (NHMRC 2007) and therefore were either pseudorandomised controlled trials or comparative studies with or without concurrent controls. Previous reviews (Law *et al.* 2003, Lee and Gibbon 2015, Morgan and Vogel 2008) have followed more restrictive criteria with regards to study design. However, in order to reflect the growing evidence base and the potential for lower graded studies to develop into larger studies with more robust research designs, the decision was made to include studies with a lower level of evidence, as defined by NHMRC (2007). This allowed an investigation of the current level of evidence for interventions and a clear picture regarding what is required to take the evidence forward. As a counter to the inclusion of studies with lower graded evidence, the quality appraisal tools were used to identify studies with the most robust operationalisations of these designs and reporting processes.

It should be noted, nevertheless, that where higher graded study designs were used, results could shed further light on lower graded designs. For example, whereas the studies by Gierut and colleagues (1989, 1990, 1996) showed a positive outcome for the complexity approach in single case designs, Rvachew and Nowak (2001) found that greater change was observed in children who received input following a developmental rather than a complexity

approach to intervention in a higher graded group study. Similarly, the group study carried out by Almost and Rosenbaum (1998) provides more convincing evidence for their combined approach to intervention in comparison to the case studies reported by others within this category of interventions.

The data extraction process revealed that many studies did not report complete data regarding dosage but where these were reported, there was a wide range in the number of sessions provided (three to 67). However, there were no clear patterns to the dosage provided within the categories and subcategories of interventions. Rather, where it was reported, a wide range of number, frequency and duration of intervention sessions were offered. A lack of consistency in the provision of intervention makes it harder to compare across interventions and to determine the relative benefit of each.

With regards to measuring outcomes, a range of tools was used to assess speech output including published assessments, picture naming tasks and spontaneous continuous speech samples. As with dosage, there were no clear patterns within the categories and subcategories with regard to outcome data collection and analysis. Thus, a narrative synthesis has been used rather than attempt a meta-analysis where the measures differed widely. The exception to this was the subcategories of imitation and drill and complexity approaches which both relied heavily on probe word lists to test outcomes. However, these studies were predominantly carried out by two groups of researchers which may explain the tendency towards the same measurement tools rather than indicating consensus across research groups in favour of any particular measure.

The model for classifications of interventions for SSD

The classification model used to classify those interventions which were included in the review was developed using a bottom-up approach based on interventions described by

clinicians in practice (Roulstone and Wren 2001). The model proposes five main categories (environmental, auditory-perceptual, cognitive-linguistic, production and integrated) that distinguish interventions according to where change, which will lead to improved speech output, is expected to occur. The subcategories attempt to capture more precisely what is being asked of the child in order to effect change. An exhaustive list of possibilities is not presented however and the model will undoubtedly evolve as new intervention procedures emerge and the evidence base grows.

Mapping the evidence to the model

Categorisation of studies to the model was complex. Many of the studies included could have been categorised under the subcategory of 'combined', for example all three of the studies listed under auditory perceptual included production activities. However, studies were categorised according to the specific element of the intervention being investigated. Some studies added components to their interventions during the course of their study making it difficult to assess the particular contribution to outcome relative to the original aim of the study (McIntosh and Dodd 2008, Saben and Ingham 1991). Further difficulties arose concerning the amount of information regarding intervention procedures provided in the paper. With more information, it is possible that some of the studies reported would be re-categorised into a different group.

The majority of studies in the review focused on just three of the eleven subcategories of the model: imitations and drill (seven studies), meaningful minimal contrasts (three studies) and complexity (six studies). The remaining studies covered a further four categories/subcategories. Thus, no studies were identified for four of the subcategories of the model. It is possible that no evidence is available for each of these subcategories or that the evidence that is available was not robust enough to be included in

the review, despite the broader inclusion criteria of this review compared to others. Rather than suggesting that those subcategories with no studies in the review are ineffective, the more accurate conclusion would be that currently, there is no strong evidence to support these intervention procedures with preschool aged children.

Some degree of supporting evidence was identified for seven of the intervention categories and subcategories in the model. These covered all of the five main categories and a range of subcategories: environmental approaches; phoneme perception; guidance on phonetic placement/manner; imitations and drill; contrasts; complexity; combined and unspecified approaches. The number of quality studies varied across these subcategories, from just one each for 'environmental' and 'guidance on phonetic placement/manner' to seven for imitation and drill. Three subcategories in the model, imitations and drill, contrasts and complexity, were supported by a number of good quality studies but the level of evidence represented in each of these studies is low based on the NHMRC classification of levels of evidence (NHRMC 2007). Across these three subcategories of intervention procedure, the highest graded study was at level III-2 – a comparative study with concurrent controls. This is comparable with a classification of indicative evidence based on the 'What Works' database of interventions (Law *et al.* 2015). The fact that there are studies with higher grade evidence adds credence to the findings for the category or subcategory as a whole but there is still a need for more studies utilising a higher level of evidence methodologies to strengthen the evidence base for these types of intervention. This fits with the findings of Baker and McLeod (2011) who commented on the need for higher levels of scientific rigour and the importance of replication research to build on the findings of lower graded studies.

Higher grade evidence was identified in the review for three studies: one using phoneme perception (Rvachew *et al* 2004), one which used a combined approach (Almost and Rosenbaum 1998); and a third where the intervention procedure was unspecified (Glogowska *et al.*, 2000). All three studies were randomised controlled trials with large sample sizes relative to most of the other studies (34, 26 and 26 respectively). Given that a range of interventions was used within these three studies, this suggests that there is agreement that a variety of approaches to intervention can be effective for children with SSD (Lancaster *et al.* 2010).

Clinical implications

The review and categorisation of the studies onto the model of interventions, as illustrated in figure 3, provides an easy reference for clinicians regarding which interventions have evidence to support them. The categories of intervention can also be mapped onto the needs of individual children. For example, where assessment has shown that a child's presenting SSD is associated with problems in auditory processing, the interventions described by Wolfe *et al* (2003) and Rvachew and colleagues (1994, 2004) could be useful. The descriptions in the individual papers regarding both the activities which were carried out and the manner of delivery, in terms of number and frequency of sessions, can assist in providing information for an evidence based service. Similarly, if assessment reveals that a child's needs appear to be in the areas of cognitive-linguistic processing or production skill, the relevant studies in each category can be used to guide the plan for intervention. Though more comparative studies need to be completed to determine the degree to which some approaches are more effective or efficient than others within categories, the ability to identify specific approaches mapped to children with specific needs is invaluable in the

clinical context when time for considering the literature to cover a broad range of presentations for SSD is limited.

Strengths and limitations of the study

The systematic review had a specific remit to look at the evidence base related to intervention for SSD with preschool children (2;00 – 5;11). Studies with 20% or more of children outside the specified age range were not included. The criteria for inclusion meant that some frequently cited papers were not included in the review. The reasons for non-inclusion were most often related to the age range of the children in the sample or a low score on the quality appraisal tools used. Some studies were also excluded because the sample used in the study included children with known concomitant difficulties such as cleft palate or hearing loss or because outcomes were not reported for speech (see Appendices 3 and 4 for excluded studies). Moreover, as the outcome measure needed to include speech output, the review did not include interventions which focused on prosodic skills or speech perception or other underlying speech processing skills unless these were included alongside a measurement of speech output.

Conclusion

To summarise, there is evidence to support certain types of intervention for preschool children with SSD and this evidence is presented in a manner which has meaning and relevance to clinicians. Whilst there are more studies to support those interventions working on imitation and drill procedures or using cognitive-linguistic approaches, the stronger evidence is linked to working on phoneme perception, combined and unspecified approaches to intervention for children in the preschool age range. It is possible of course

that evidence for interventions may vary in older children. Given the variation in findings across different study designs, it is important nevertheless for individual clinicians to read the papers themselves to understand how the intervention was delivered, the detailed characteristics of the children for whom the intervention was effective and what specifically was being investigated.

The work so far has been invaluable in establishing a preliminary evidence base in which different intervention types have been trialled and explored through small scale studies. As well as providing initial evidence, these studies have enabled researchers to explore the facets of a particular approach to intervention. It has allowed for the understanding of issues relating to delivery which can inform both clinical practice and further investigations. There is a need now for research activity to advance the knowledge base through the use of higher graded methodological studies which will provide more robust information on which approaches or combination of approaches are most suitable to use with this client group.

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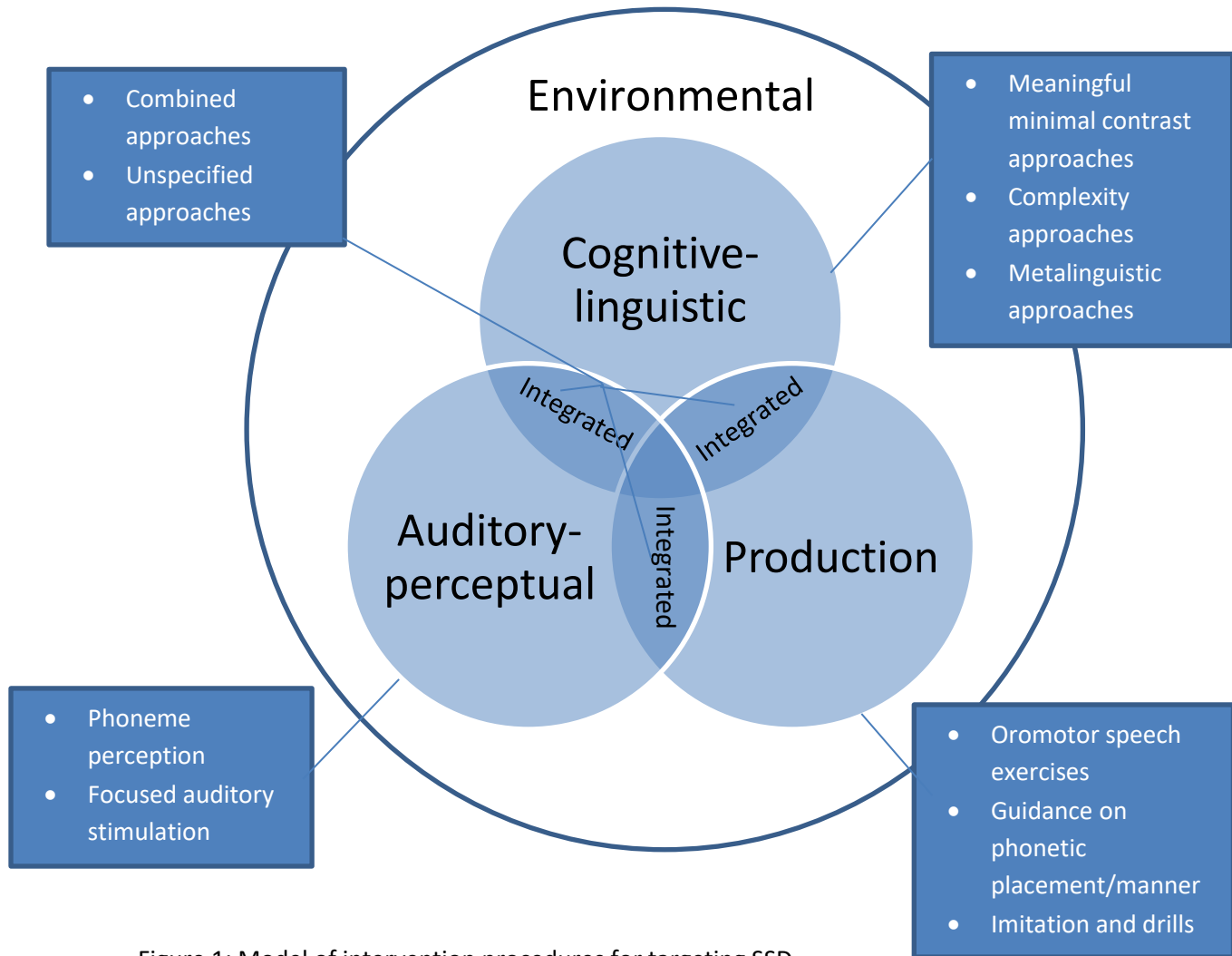


Figure 1: Model of intervention procedures for targeting SSD

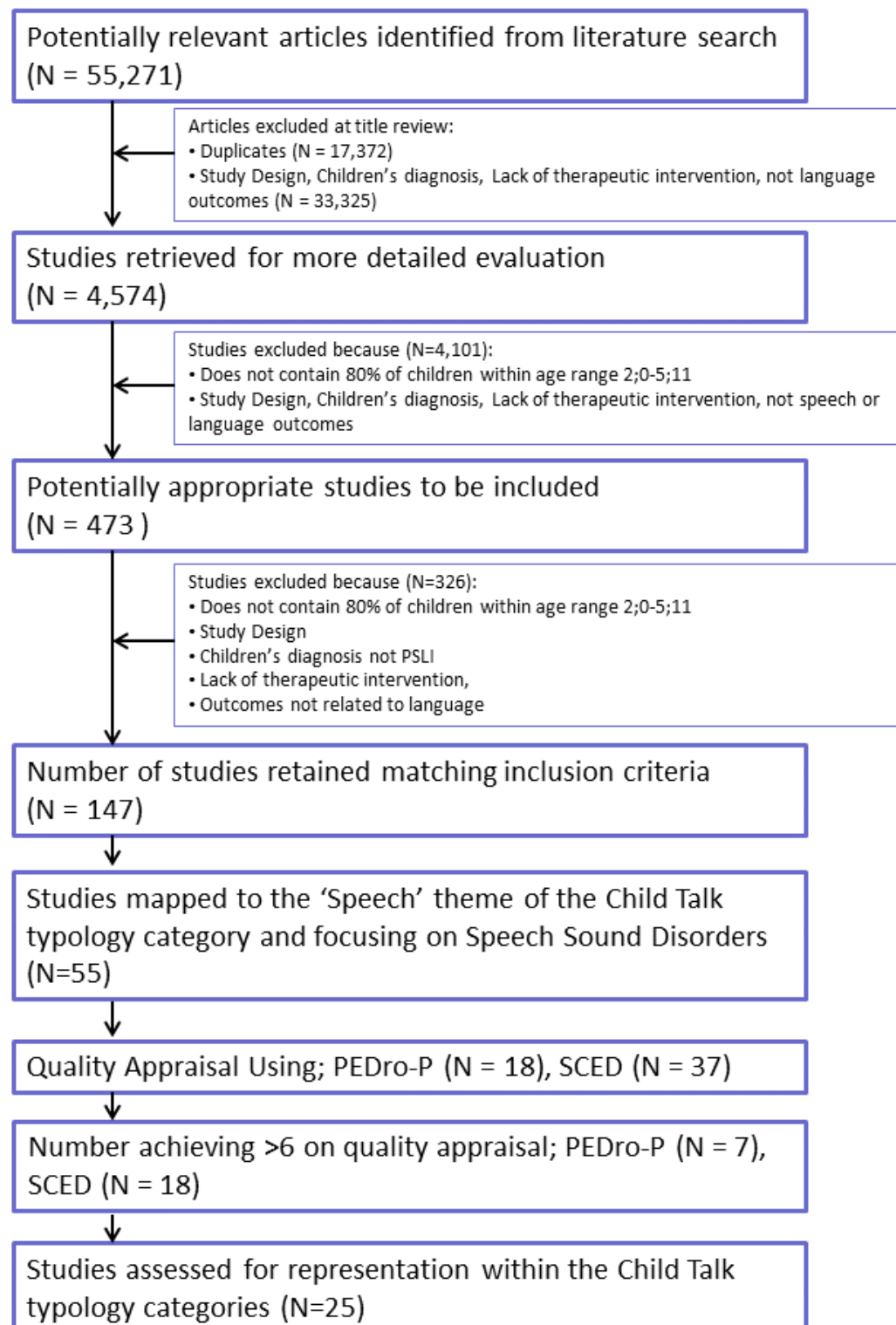
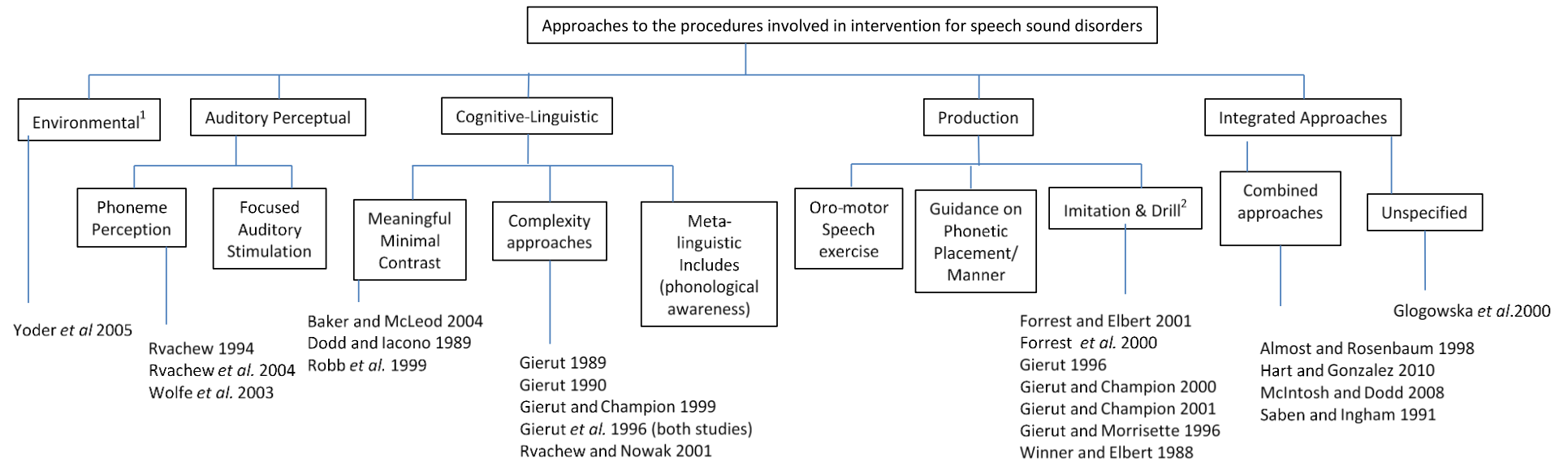


Figure 2: PRISMA flow chart



1 = Includes modelling and recasting and other approaches which are embedded in everyday interactions

2 = Imitation /spontaneous production of sounds in a progressively more complex context - syllables, words or non-words

Figure 3: Evidence for intervention procedures for preschool children with SSD

Table 1: Process of categorisation of procedures in intervention for SSD

| | Environmental | Auditory perceptual | Cognitive linguistic | Production | Combined |
|-------------|---|---|---|---|--|
| Description | Procedures which are incorporated into everyday interactions. | Procedures which are target listening and perceptual skills | Procedures which require the child to reflect on their speech and/or increase awareness of speech generally | Procedures which aim to effect change through instruction on production and production practice | Procedures which combine two or more of the other four categories into a tested intervention |
| Examples | Modelling, recasting | Auditory discrimination, focused auditory stimulation, phoneme perception tasks | Contrast therapy, metalinguistic tasks | Drills, guidance on phonetic placement or manner, traditional articulation | Cycles approach, psycholinguistic approach |

Table 2: Summary of studies from systematic review

| Study Author(s) Country of origin | No of child participants (number of children in each group, if applicable) | Age range (months) | Study Design (Type of Evidence [†]) | No. of therapy sessions/ Agent of Delivery | Length of each session | Frequency of sessions | Duration of intervention | Type of speech sampled | Analysis used to measure change | PEDro-P/SCED score | Effect Size Cohen d unless otherwise specified |
|--|---|--|--|---|------------------------|-----------------------|--------------------------|---|---|--------------------|--|
| Environmental | | | | | | | | | | | |
| Yoder, P., Camarata, S., & Gardner, E. (2005). USA | 52 (26, 26) | Group 1 – average 44.3 Group 2 – average 43.2 | Randomised (Type II) | Group 1 – Control 0; Group 2 (treatment group) 72/SLP | 30 minutes | Three times per week | 6 months | Spontaneous speech | Percentage intelligible utterance PVC* PCC* | PEDro-P 7 | 49 (taken directly from article) |
| Auditory Perceptual: Phoneme Perception | | | | | | | | | | | |
| Rvachew, S. (1994). Canada | 27 (10, 9, 8) | Group 1 – average 53.4 Group 2 – average 53.6 Group 3 – average 51.5 | Randomised (Type II) | 6 / SLP | 45 minutes | Weekly | 6 – 11 weeks | Word identification Single word naming | Percentage correct word identification Number of single words produced correctly | PEDro-P 6 | 0.0092 |
| Rvachew, S., Nowak, M., & | 34 (17, 17) | Group 1 – average 52.88 | Randomised (Type II) | 16 (in addition to their regular | 15 minutes | Weekly | 4.73 months | Conversation | PCC* | PEDro-P 6 | 0.8316 |

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| | | | | | | | | | | | |
|--|----------|--|---|------------------------|---------------|--------------|-----------------------------|----------------------------------|--|-----------|---------------------|
| Cloutier, G. (2004). Canada | | Group 2 – average 50.29 | | therapy)/ SLP | | | | | | | |
| Wolfe, V., Presley, C., & Mesaris, J. (2003). USA | 9 (4, 5) | Group 1 – 47 – 55 Group 2 – 41 – 50 | Comparative studies – Randomised Therapy approach (Type II) | Average 11 / SLP | 30 minutes | Twice weekly | One academic quarter | Probe list | Accuracy of production Sound identification | PEDro-P 6 | -0.3634 |
| Cognitive-Linguistic: Meaningful Minimal Contrast | | | | | | | | | | | |
| Baker, E., & McLeod, S. (2004). Australia | 2 | Subject 1 – 57 Subject 2 – 52 | Single Subject studies – Case Report, A-B, Multiple Baseline Design (Type IV) | 1 – 12 2 – 32 / SLP | 45 minutes | Twice weekly | 1 – 6 weeks 2 – 16 weeks | Probe conversation | Percentage correct production of trained cluster | SCED 7 | 0.001 [‡] |
| Dodd, B., & Iacono, T. (1989). Australia | 7 | 36 – 57 | Case Series Pre-Post Intervention Design (Type IV) | 3 – 40 / SLP | Not available | Weekly | Average 23.6 weeks | Spontaneous speech (during play) | PCC* Phoneme Inventory Process analysis RIU* | SCED 6 | -1.362 [‡] |
| Robb, M. P., Bleile, K. M., & Yee, S. S. L. (1999). USA | 1 | 48 | Case study - Single Subject Pre-Post Intervention Design (Type IV) | 20 / SLP | 45 minutes | Twice weekly | 10 weeks | Speech sample Probe list | Percentage accuracy Vowel inventory PVC* Acoustic analyses of vowels (duration, fundamental frequency) | SCED 6 | Insufficient data |
| Cognitive-Linguistic: Complexity Approaches | | | | | | | | | | | |

RUNNING HEAD: Systematic review of speech interventions

| | | | | | | | | | | | |
|--|---|---------|--|---------------------|---------------|----------------------|-----------------------|-------------|---|--------|--|
| Gierut, J. A. (1989). USA | 1 | 55 | Case Study, Pre-Post Intervention Design (Type IV) | 23 / SLP | 30 minutes | Twice weekly | 11.5 weeks | Probe lists | Percentage accurate production of target phonemes | SCED 8 | Insufficient data |
| Gierut, J. A. (1990). USA | 3 | 49 – 58 | Alternating treatment design – Multiple Baseline Design (Type III-3) | Not available / SLP | 60 minutes | Three times a week | Not available | Probe list | Percentage accuracy correct on probe list | SCED 9 | Figures are of insufficient resolution to extract data |
| Gierut, J. A., & Champion, A. H. (1999). USA | 2 | 48 – 56 | Single Subject studies – Multiple Baseline Design (Type III-3) | 12 / SLP | 60 minutes | Three times per week | Approximately 7 weeks | Probe | Percentage accuracy correct on probe list | SCED 6 | Figures are of insufficient resolution to extract data |
| Gierut, J. A., Morrisette, M. L., Hughes, M. T., & Rowland, S. (1996). STUDY 1 USA | 3 | 43 – 66 | Single Subject studies – alternating treatment design (Type III-2) | Up to 19 / SLP | 60 minutes | Three times per week | Not available | Probe list | Percentage accuracy correct on probe list | SCED 7 | Figures are of insufficient resolution to extract data |
| Gierut, J. A., Morrisette, M. L., | 6 | 41 – 66 | Single Subject studies – Multiple Baseline Design | Not available / SLP | Not available | Not available | Not available | Probe list | Percentage accuracy correct on probe list | SCED 7 | Figures are of insufficient |

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|--|-------------|--|----------------------|----------|---------------|--------|-----------------------------|-----------------------|---------------|-----------|----------------------------|
| Hughes, M. T., & Rowland, S. (1996). STUDY 2 USA | | | (Type III-2) | | | | | | | | resolution to extract data |
| Rvachew, S., & Nowak, M. (2001). Cnada | 48 (24, 24) | Group 1 – average 51.46 Group 2 – average 49.63 | Randomised (Type II) | 12 / SLP | Not available | Weekly | 12 weeks in two blocks of 6 | PPKP* Conversation | PPKP* PCC* | PEDro-P 6 | -0.1194 |

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| Production: Imitation & drills | | | | | | | | | | | |
|---|-----------|---------|--|---------------------|---------------|----------------------|------------------|------------|---|--------|--------------------------------------|
| Forrest, K., & Elbert, M. (2001). USA | 4 | 59 – 63 | Single Subject studies – Multiple Baseline Design (Type III-2) | Not available / SLP | 45 minutes | Twice weekly | Not available | Probe list | PCC* for target phonemes | SCED 6 | Insufficient data |
| Forrest, K., Elbert, M., & Dinnsen, D. A. (2000). USA | 10 (5, 5) | 40 – 54 | Comparative studies – Therapy approach (Type III-3) | Not available / SLP | Not available | Fortnightly | Not available | Probe | Percentage accuracy correct of probe | SCED 8 | Insufficient data |
| Gierut, J. A. (1996). USA | 7 | 40 – 68 | Single Subject studies – Multiple Baseline Design (Type III-2) | Not available / SLP | 60 minutes | Three time per week | Average 18 weeks | Probe | Change in phonemic inventory | SCED 6 | Insufficient data |
| Gierut, J. A., & Champion, A. H. (2000). USA | 1 | 53 | Single Subject studies – Multiple Baseline Design (Type III-2) | 19 / SLP | 60 minutes | Three times per week | 19 | Probe list | Percentage accuracy correct on probe list | SCED 6 | Insufficient data |
| Gierut, J. A., & Champion, A. H. (2001). USA | 8 | 40 – 75 | Single Subject studies – Multiple Baseline Design (Type III-2) | Not available / SLP | 60 minutes | Three times per week | Not available | Probe list | Percentage accuracy correct on probe list | SCED 9 | IRD ^π - between 84 & 100% |

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|--|-------------|---------|--|-------------------------|---------------|----------------------|---|---|--|-----------|--------------------------------------|
| Gierut, J. A., & Morrisette, M. L. (1996). USA | 2 | 47 – 62 | Single Subject studies – Multiple Baseline Design (Type III-2) | Not available / SLP | 60 minutes | Three time per week | Average of 16 weeks | Probes | Phoneme inventory | SCED 6 | Insufficient data |
| Winner, M., & Elbert, M. (1988). USA | 4 | 46 – 68 | Single subject studies – Multiple Baseline Design (Type III-2) | 25 / SLP | 30 minutes | Three times per week | 8 weeks | Speech sample Probe list Spontaneous speech (picture description) | Percentage correct scores of target sounds | SCED 7 | IRD ^π - Between 50 & 100% |
| Integrated Approaches: Combined | | | | | | | | | | | |
| Almost, D., & Rosenbaum, P. (1998). Canada | 26 (13, 13) | 33 – 61 | Group studies - Randomised (Type II) | 14 – 29 / SLP | 30 minutes | Twice weekly | 7 – 15 weeks | GFTA* APP-R* Standardised test of single words Conversational speech | Single words No of errors PCC* | PEDro-P 9 | 0.0004 |
| Hart, S., & Gonzalez, L. (2010). USA | 3 | 43 – 59 | Single Subject studies – Multiple Baseline Design (Type III-2) | 12 / SLP | 30 minutes | Twice a week | 6 weeks | HAPP-R 3* Spontaneous speech sample | Process analysis Percentage sample correct | SCED 8 | IRD ^π - between 0 & 100% |
| McIntosh, B., & Dodd, B. (2008). Australia | 3 | 36 - 45 | Single Subject Pre-Post Intervention Design (Type IV) | Between 12 and 38 / SLP | 30-40 minutes | Twice Weekly | Between 6 and 19 weeks (average 12.8 weeks) | Single word naming test (DEAP* phonology subtest) | PVC* PCC* PPC* Percentage inconsistency | SCED 6 | -42.187 [¥] |

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| | | | | | | | | | | | |
|--|--------------|--|--|-------------------------|-----------------------|---------------|-------------------------------|---|--|-----------|-------------------|
| | | | | | | | | Connected speech task (DEAP*) Repeated production of words (DEAP* – inconsistency subtest) | | | |
| Saben, C. B., F & Ingham, J. C. (1991). USA | 2 | Subject 1 -52 Subject 2 – 45 | Single Subject studies – Multiple Baseline Design (Type III-2) | 1 – 67 2 – 32 / SLP | Not available | Not available | 1 – 9 months 2 – 4½ months | Probe list (spontaneous picture naming) | Percentage use of individual targeted phonemic processes | SCED 8 | Insufficient data |
| Integrated Approaches: Unspecified | | | | | | | | | | | |
| Glogowska, M., Roulstone, S., Enderby, P., & Peters, T. J. (2000). UK | 159 (71, 84) | Group 1 – 18 – 42 Group 2 – 24 – 42 | Comparative studies – Randomised Therapy approach (Type II) | Average 6.2 hours / SLP | Average of 47 minutes | Once a month | Average of 8.4 months | Unclear | Error rate | PEDro-P 8 | 0.0477 |

[†]NHMRC (2007) Evidence Hierarchy: Designations of 'levels of evidence' according to type of research question.

*APP-R: the Assessment of Phonological Processes – Revised (Hodson, 1986); DEAP: Diagnostic Evaluation of Articulation and Phonology, (Dodd, Zhu, Crosbie, Holm and Ozanne 2002); GFTA: Goldman Fristoe Test of Articulation (Goldman and Fristoe 1969, 2000); HAPP-R: the Assessment of Phonological Processes – Revised (Hodson 2004); PCC – Percent Consonants Correct; PPC – Percent Phonemes Correct; PVC: percentage vowels correct; (Shriberg and Kwiatkowski 1982); PPKP – Productive Phonological Knowledge Profile (Gierut, Elbert and Dinnsen 1987); Psycholinguistic Framework (Stackhouse and Wells 1997); RIU – Relative Influence on Unintelligibility (Dodd and Iacono, 1989).

¥Effect size calculated using a within subject design and online calculator from <http://www.cognitiveflexibility.org/effectsize/effectsizecalculator.php>

Π- IRD =Improvement Rate Difference – a method of calculating effect size for single-subject experimental designs (Parker, Vannest and Davis, 2011)

Appendix 1: Search terms used in systematic review of interventions for SSD in preschool children

1. exp Pediatrics/
2. exp CHILD/
3. exp INFANT/
4. child\$.mp. [mp=title, original title, abstract, name of substance word, subject heading word]
5. infant\$.mp. [mp=title, original title, abstract, name of substance word, subject heading word]
6. (paediatric\$ or pediatric\$).mp. [mp=title, original title, abstract, name of substance word, subject heading word]
7. toddler\$.mp. [mp=title, original title, abstract, name of substance word, subject heading word]
8. boy\$.ti,ab.
9. girl\$.ti,ab.
10. (school child\$ or schoolchildren\$).ti,ab.
11. (pre school\$ or preschool\$).ti,ab.
12. or/1-11
13. speech disorder\$.ti,ab.
14. speech intelligibility\$.ti,ab.
15. speech therap\$.ti,ab.
16. language therap\$.ti,ab.
17. speech development.ti,ab.
18. speech delay.ti,ab.
19. language disorder\$.ti,ab.
20. language development disorder\$.ti,ab.
21. sign language\$.ti,ab.
22. child\$ language.ti,ab.
23. language therap\$.ti,ab.
24. language development.ti,ab.
25. language delay.ti,ab.
26. nonverbal communication.ti,ab.
27. non verbal communication.ti,ab.
28. communication development.ti,ab.
29. exp Speech Disorders/
30. speech Intelligibility/
31. "rehabilitation of speech and language disorders"/ or language therapy/ or speech therapy/
32. Language Development Disorders/
33. Language Disorders/
34. Sign Language/
35. Child Language/
36. Language Development/
37. exp Nonverbal Communication/
38. Communication Disorders/
39. maternal responsiveness.tw.
40. directiveness.tw.
41. maternal interactive styles.tw.
42. compliance.tw.
43. maternal personality.tw.
44. child temperament.tw.
45. or/13-44
46. exp Mental Retardation/
47. exp child development disorders, pervasive/ or asperger syndrome/
48. Cleft Palate/ or Cleft Lip/
49. Otitis Media with Effusion/
50. exp Hearing Loss/
51. exp Blindness/
52. Stuttering/

53. Aphonia/
54. exp Pain/
55. Crying/
56. exp Analgesia/
57. Reading/
58. exp Dyslexia/
59. Cerebral Palsy/
60. (alternative and augmentative communication).mp. [mp=title, original title, abstract, name of substance word, subject heading word]
61. "alternative and augmentative communication".mp. [mp=title, original title, abstract, name of substance word, subject heading word]
62. exp aged/
63. geriatrics/
64. or/46-63
65. (12 and 45) not 64
66. randomized controlled trial.pt.
67. controlled clinical trial.pt.
68. randomized controlled trials/
69. random allocation/
70. double blind method/
71. single blind method/
72. clinical trial.pt.
73. exp clinical trials/
74. (clin\$ adj25 trial\$).tw.
75. ((singl\$ or doubl\$ or trebl\$ or tripl\$) adj25 (blind\$ or mask\$)).tw.
76. placebos/
77. placebo\$.tw.
78. random\$.tw.
79. research design/
80. "comparative study"/
81. exp evaluation studies/
82. follow-up studies/
83. prospective studies/
84. (control\$ or prospectiv\$ or volunteer\$).tw.
85. (control\$ or prospectiv\$ or volunteer\$).tw.
86. or/66-85
87. "animal"/
88. "human"/
89. 87 not 88
90. 86 not 89

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Appendix 2: Databases searched, number of results and search date

| Database* | Search interface | Search results | Search date |
|---|----------------------|----------------|------------------|
| MEDLINE | Ovid | 8374 | 6 December 2011 |
| EMBASE | Ovid | 9663 | 6 December 2011 |
| Cumulative Index to Nursing and Allied Health Literature (CINAHL) | EBSCOhost | 8976 | 2 December 2011 |
| PsycINFO | EBSCOhost | 9107 | 11 January 2011 |
| Cochrane Database of Systematic Reviews (CDSR) | The Cochrane Library | 255 | 13 January 2012 |
| Database of Abstracts of Reviews of Effects (DARE) | The Cochrane Library | 0 | 13 January 2012 |
| NHS Health Technology Assessment database | The Cochrane Library | 0 | 13 January 2012 |
| Cochrane Central Register of Controlled Trials (CENTRAL) | The Cochrane Library | 0 | 13 January 2012 |
| Science Citation Index | Web of Knowledge | 5787 | 13 January 2012 |
| Social Science Citation Index | Web of Knowledge | 0 | 13 January 2012 |
| International Bibliography for the Social Sciences | ProQuest | 0 | 25 November 2011 |
| Applied Social Sciences Index and Abstracts (ASSIA) | ProQuest | 1799 | 25 November 2011 |
| Sociological Abstracts ProQuest 3800 25 November 2011 | ProQuest | 3800 | 25 November 2011 |
| Social Services Abstracts ProQuest 0 25 November 2011 | ProQuest | 0 | 25 November 2011 |
| Educational Resource Information Center (ERIC) | ProQuest | 4000 | 26 January 2012 |
| Linguistics and Language Behavior Abstracts | ProQuest | 3006 | 20 January 2012 |
| British Education Index | ProQuest | 464 | 20 January 2012 |

| | | | |
|---|--|----|-----------------|
| The Campbell Collaboration | www.campbellcollaboration.org/ | 40 | 13 January 2012 |
| *Databases were searched from the date of inception up to the search date | | | |

| Reference | Reason for exclusion |
|---|--|
| Alant, E. and Jager, G. "Integrated speech and language therapy in the pre-school class in the special school setting." <i>Rehabilitation in South Africa</i> 27.2 (1983): 49-53. | Description of project. Not a research article |
| Aram, D. M., Morris, R. and Hall, N.E. "The Validity of Discrepancy Criteria for Identifying Children with Developmental Language Disorders." <i>Journal of learning disabilities</i> 25.9 (1992): 549-54. | Observational study |
| Baker, E. "The experience of discharging children from phonological intervention." <i>International Journal of Speech language Pathology</i> 12.4 (2010): 325-28. | Discussion article |
| Baker, E. "Management of speech impairment in children: The journey so far and the road ahead." <i>Advances in Speech Language Pathology</i> 8.3 (2006): 156-63. | Discussion article |
| Baker, E. and McCabe, P. "The Potential Contribution of Communication Breakdown & Repair in Phonological Intervention." <i>Canadian Journal of Speech-Language Pathology and Audiology/Revue canadienne d'orthophonie et d'audiologie</i> 34.3 (2010): 193-205. | Review and Discussion article |
| Befi-Lopes, D. M. and Rondon, S. "Syllable Deletion in Spontaneous Speech of Children with Specific Language Impairment." <i>PRO-FONO: Revista de Actualizacao Cientifica</i> 22.3 (2010): 333-38. | Observational study |
| Bernhardt, B. H., Stemberger, J.P., and Major, E. "General and nonlinear phonological intervention perspectives for a child with a resistant phonological impairment." <i>Advances in Speech Language Pathology</i> 8.3 (2006): 190-206. | Discussion article |
| Blacklin, J. and Crais, E.R.. "A treatment protocol for young children at risk for severe expressive output disorders." <i>Seminars in Speech & Language</i> 18.3 (1997): 213. | Discussion article |
| Bland, L. E. and Prelock, P. A. "Effects of Collaboration on Language Performance." <i>Communication Disorders Quarterly</i> 17.2 (1995): 31-37. | Participants too old |
| Bowen, C. and Cupples, L. "The role of families in optimizing phonological therapy outcomes." <i>Child Language Teaching and Therapy</i> 20.3 (2004): 245-60. | Discussion article |

| Reference | Reason for exclusion |
|--|--|
| Boyle, J. "Speech and language delays in preschool children." British medical journal 343 (2011): d5181. | Editorial |
| Browning, E. "The health visitor and speech impaired children." Health visitor 54 (1981): 204-05. | Discussion article |
| Carter, P. and S. Edwards. "EPG therapy for children with longstanding speech disorders: predictions and outcomes." Clinical Linguistics & Phonetics 18.6 (2004): 359-72. | Participants too old |
| Castiglia, P. T. "Speech-language development." Journal of Pediatric Healthcare 1.3 (1987): 165-67. | Discussion article |
| Chang, J. Y. "Case study on a profound speech-delayed subject: a behavioural approach and its implications." Asia Pacific Journal of Speech, Language & Hearing 9.1 (2004): 48-53. | Could not exclude language delay as part of other developmental or biological disorder |
| Cox, J. and Hill, S. "Tackling language delay: a group work approach." Health visitor 66.8 (1993): 291-92. | Discussion article |
| Daly, D. A., Cantrell, R.P. Cantrell, M.L. and Aman, L.A. "Structuring speech therapy contingencies with an oral apraxic child." The Journal of speech and hearing disorders 37.1 (1972): 22-32. | Participant too old |
| Danger, S. and Landreth, G. "Child-Centered Group Play Therapy with Children with Speech Difficulties." International Journal of Play Therapy 14.1 (2005): 81-102. | Unable to establish if participants are an appropriate age |
| Denne, M., Langdown, N., Pring, T. and Roy, P. "Treating children with expressive phonological disorders: does phonological awareness therapy work in the clinic?" International Journal of Language & Communication Disorders 40.4 (2005): 493-504. | Greater than 20% of the participants were too old |
| Dodd, B., Crosbie, S., McIntosh, B., Holm, A., Harvey, C., Liddy, M., Fontyne, K., Pinchin, B. and Rigby, H. "The impact of selecting different contrasts in phonological therapy." International Journal of Speech-Language Pathology 10.5 (2008): 334-45. Forrest, K. and Iuzzini, J. "A comparison of oral motor and production training for children with speech sound disorders." Seminars in Speech & Language 29.4 (2008): 304-11. | Greater than 20% of the participants were too old |

| Reference | Reason for exclusion |
|---|---|
| Forrest, K. and Iuzzini, J. "A comparison of oral motor and production training for children with speech sound disorders." <u>Seminars in Speech & Language</u> 29.4 (2008): 304-11. | Greater than 20% of the participants were too old |
| Gierut, J. A. "Natural domains of cyclicity in phonological acquisition." <u>Clinical Linguistics & Phonetics</u> 12.6 (1998): 481-99. | Age unclear, greater than 20% likely to be too old. Outcome measure phonology only |
| Gierut, J. A., Morrisette, M.L. and Ziemer, S.M. "Nonwords and generalization in children with phonological disorders." <u>American Journal of Speech-Language Pathology</u> 19.2 (2010): 167-77. | Greater than 20% of the participants were too old |
| Gillon, G. T. "The efficacy of phonological awareness intervention for children with spoken language impairment." <u>Language Speech and Hearing Services in Schools</u> 31.2 (2000): 126-41. | Greater than 20% of the participants were too old |
| Gillon, G. T. "Follow-up study investigating the benefits of phonological awareness intervention for children with spoken language impairment." <u>International Journal of Language & Communication Disorders</u> 37.4 (2002): 381-400. | Greater than 20% of the participants were too old |
| Keske-Soares, M., Brancalioni, A.R., Marini, C., Pagliarin, K.C. and Ceron, M.I. "Therapy effectiveness for phonological disorders with different therapeutic approaches." <u>Pro-Fono</u> 20.3 (2008): 153-58. | Greater than 20% of the participants were too old |
| King, A. M. "An Integrated Multimodal Intervention Approach to Support Speech and Language Development in Children With Severe Speech Impairments." Diss. ProQuest Information & Learning, 2011. | Greater than 20% of the participants were too old |
| Lane, K. L., Fletcher, T., Carter, E.W., Dejud, C. and DeLorenzo, J. "Paraprofessional-led phonological awareness training with youngsters at risk for reading and behavioral concerns." <u>Remedial & Special Education</u> 28.5 (2007): 266-76. | Participants were too old |
| Mire, S.P. and Montgomery, J.K. "Early Intervening for Students With Speech Sound Disorders. Lessons From a School District." <u>Communication Disorders Quarterly</u> 30.3 (2009): 155-66. | Review article |
| Pascoe, M., Stackhouse, J. and Wells, B. "Phonological therapy within a psycholinguistic framework: promoting change in a | Case study - Child was too old |

| Reference | Reason for exclusion |
|---|---|
| child with persisting speech difficulties." International Journal of Language & Communication Disorders 40.2 (2005): 189-220. | |
| Robertson, S. B. and Weismer, S.E. "Effects of treatment on linguistic and social skills in toddlers with delayed language development." Journal of Speech, Language & Hearing Research 42.5 (1999): 1234-48. | Greater than 20% of the participants were too young |
| Roulstone, S., Glogowska, M., Peters, T.J. and Enderby, P. "Building good practice: lessons from a multimethod study of speech and language therapy... including commentary by Tyler AA." International Journal of Therapy & Rehabilitation 11.5 (2004): 199-205. | Discussion article |
| Rousseau, I. Packman, A., Onslow, M., Harrison, E. Jones, M. "An investigation of language and phonological development and the responsiveness of preschool age children to the Lidcombe Program." Journal of communication disorders 40.5 (2007): 382-97. | Stuttering |
| Ruder, K. F. and Bunce, B.H.. "Articulation Therapy Using Distinctive Feature Analysis to Structure the Training Program: Two Case Studies." Journal of Speech and Hearing Disorders 46.1 (1981): 59-65. | Review paper with case study without relevant assessments |
| Ruscello, D. M. "Visual feedback in treatment of residual phonological disorders." Journal of communication disorders 28.4 (1995): 279. | Discussion article |
| Ruscello, D. M., Yanero, D. and Ghalichebaf, M. "Cooperative service delivery between a university clinic and a school system." Language, Speech & Hearing Services in Schools 26.3 (1995): 273-77. | Case study child too old |
| Segers, E., Nooijen, M. and de Moor, J. "Computer vocabulary training in kindergarten children with special needs." International Journal of Rehabilitation Research 29.4 (2006): 343-45. | Greater than 20% of the participants were too old |
| Segers, E. and Verhoeven, L. "Effects of lengthening the speech signal on auditory word discrimination in kindergartners with SLI." Journal of communication disorders 38.6 (2005): 499-514. | Greater than 20% of the participants were too old |

| Reference | Reason for exclusion |
|--|---|
| Sharp, H. M. and Hillenbrand, K. "Speech and Language Development and Disorders in Children." <i>Pediatric clinics of North America</i> 55.5 (2008): 1159-+. | Discussion article |
| Shiller, D. M., Rvachew, S. and Brosseau-Lapre, F. "Importance of the Auditory Perceptual Target to the Achievement of Speech Production Accuracy." <i>Canadian Journal of Speech-Language Pathology and Audiology/Revue canadienne d'orthophonie et d'audiologie</i> 34.3 (2010): 181-92. | Case study 1 - child too old Case study 2 - English was a second language |
| Simser, J. I. "Auditory-verbal intervention: infants and toddlers." <i>Volta Review</i> 95.3 (1993): 217-29. | Discussion article |
| Skau, L. and Cascella, P.W. "Using Assistive Technology to Foster Speech and Language Skills at Home and in Preschool." <i>TEACHING Exceptional Children</i> 38.6 (2006). | Discussion article |
| Skelton, S. L. "Concurrent task sequencing in single-phoneme phonologic treatment and generalization." <i>Journal of communication disorders</i> 37.2 (2004): 131-55. | Children were too old |
| Skelton, S. L. and Funk, T.E. "Teaching speech sounds to young children using randomly ordered, variably complex task sequences." <i>Perceptual and motor skills</i> 99.2 (2004): 602-04. | Can not exclude the possibility that impairment is due to other developmental disorder or pervasive condition |
| Stackhouse, J., Wells, B., Pascoe, M. and Rees, R. "From phonological therapy to phonological awareness." <i>Seminars in Speech & Language</i> 23.1 (2002): 27. | Review article |
| Torgesen, J. K. and Davis, C. "Individual difference variables that predict response to training in phonological awareness." <i>Journal of experimental child psychology</i> 63.1 (1996): 1-21. | Children were 'at risk' of impairment |
| Tyler, A. A. "Language-based intervention for phonological disorders." <i>Seminars in speech and language</i> 23.1 (2002): 69-81. | Review article with case study example |
| Tyler, A. A. "What works: evidence-based intervention for children with speech sound disorders." <i>Seminars in Speech & Language</i> 29.4 (2008): 320-30. | Review article |
| Tyler, A. A., Lewis, K.E., Haskill, A., and Tolbert, L.C. "Clinical forum. Efficacy and cross-domain effects of a morphosyntax | No speech measures |

| Reference | Reason for exclusion |
|---|---|
| and a phonology intervention." <i>Language, Speech & Hearing Services in Schools</i> 33.1 (2002): 52-66. | |
| Vicsi, K., Kovacs-Vass, E. and Barczikay, P. "A Speech Improvement Technique Based on Visual Feedback." <i>Acta Linguistica Hungarica</i> 42.1-2 (1994): 93-101. | Review article |
| Warrick, N. and Rubin, H. "Phonological Awareness: Normally Developing and Language Delayed Children." <i>Journal of Speech-Language Pathology and Audiology/Revue d'orthophonie et d'audiologie</i> 16.1 (1992): 11-20. | Only phonological awareness measures |
| Willems, S.G., Lombardino, L.L., MacDonald, J.D., and Owens, R.E. "Total communication: Clinical report on a parent-based language training program." <i>Education & Training of the Mentally Retarded</i> 17.4 (1982): 293-98. | Greater than 20% of the participants were too old |
| Williams, A. L. "Assessment, target selection, and intervention: dynamic interactions within a systemic perspective." <i>Topics in Language Disorders</i> 25.3 (2005): 231-242. | Greater than 20% of the participants were too old |
| Williams, A. L. "Clinical focus. Multiple oppositions: case studies of variables in phonological intervention." <i>American Journal of Speech-Language Pathology</i> 9.4 (2000): 289-99. | Greater than 20% of the participants were too old |
| Wren, Y. and S. Roulstone. "A comparison between computer and tabletop delivery of phonology therapy." <i>International Journal of Speech-Language Pathology</i> 10.5 (2008): 346-63. | Greater than 20% of the participants were too old |
| Zhang, X. and J. B. Tomblin. "The association of intervention receipt with speech-language profiles and social-demographic variables." <i>American Journal of Speech-Language Pathology</i> 9.4 (2000): 345-57. | Greater than 20% of the participants were too old |

Appendix 3: Studies excluded at quality appraisal phase

- Adams, C., Nightingale, C., Hesketh, A., & Hall, R. (2000). Targeting metaphonological ability in intervention for children with developmental phonological disorders. *Child Language Teaching and Therapy*, 16, 285-299.
- Baker, E. & Bernhardt, B. (2004). From hindsight to foresight: working around barriers to success in phonological intervention. *Child Language Teaching & Therapy*, 20, 287-318.
- Bowen, C. & Cupples, L. (1998). A tested phonological therapy in practice. *Child Language Teaching & Therapy*, 14, 29-50.
- Bryan, A. & Howard, D. (1992). Frozen phonology thawed: the analysis and remediation of a developmental disorder of real word phonology. *European Journal of Disorders of Communication*, 27, 343-365.
- Dodd, B. & Barker, R. (1990). The Efficacy of Utilizing Parents and Teachers as Agents of Therapy for Children with Phonological Disorders. *Australian Journal of Human Communication Disorders*, 18, 29-45.
- Dodd, B. & Barker, R. (1990). The Efficacy of Utilizing Parents and Teachers as Agents of Therapy for Children with Phonological Disorders. *Australian Journal of Human Communication Disorders*, 18, 29-45.
- Eiserman, W. D., Weber, C., & McCoun, M. (1995). Parent and Professional Roles in Early Intervention. A Longitudinal Comparison of the Effects of two Intervention Configurations. *Journal of Special Education*, 29, 20-44.
- Fey, M. E. (1993). Two Approaches to the Facilitation of Grammar in Children with Language Impairment: An Experimental Evaluation. *Journal of speech and hearing research*, 36, 141-157
- Fey, M. E., Cleave, P. L., & Long, S. H. (1997). Two models of grammar facilitation in children with language impairments: phase 2. *Journal of Speech, Language & Hearing Research*, 40, 5-19.
- Fey, M. E., Cleave, P. L., Ravida, A. I., Long, S. H., Dejmaj, A. E., & Easton, D. L. (1994). Effects of grammar facilitation on the phonological performance of children with speech and language impairments. *Journal of Speech & Hearing Research*, 37, 594-607
- Gierut, J. A. (1998a). Production, conceptualization and change in distinctive featural categories. *Journal of child language*, 25, 321-341.

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